

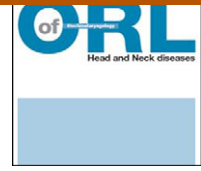


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## UPDATE

# Management of laryngomalacia

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## KEYWORDS

Stridor;  
Laryngomalacia;  
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Laser;  
Supraglottoplasty

**Summary** Laryngomalacia is the most common laryngeal disease of infancy. It is poorly tolerated in 10% of cases, requiring assessment and management, generally surgical. Surgery often consists of supraglottoplasty, for which a large number of technical variants have been described. This surgery, performed in an appropriate setting, relieves the symptoms in the great majority of cases with low morbidity. However, few data are available concerning the objective results: preoperative and postoperative objective assessment of these infants is therefore necessary whenever possible. Noninvasive ventilation (NIV) may be indicated in some infants with comorbid conditions or failing to respond to surgical management.

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Laryngomalacia is defined as collapse of supraglottic structures during inspiration. Most forms of laryngomalacia are minor (70–90%), presenting in the form of isolated and intermittent stridor with no alteration of crying or coughing, no dyspnoea, and no swallowing disorders. These minor forms do not have any consequences on the infant's growth and simply require surveillance by the paediatrician or general practitioner to detect any signs of severity.

Only severe forms of laryngomalacia require therapeutic intervention.

Signs of severity are:

- poor weight gain (probably the most contributive element);
- dyspnoea with permanent and severe intercostal or xiphoid retraction;
- episodes of respiratory distress;
- obstructive sleep apnoea;
- episodes of suffocation while feeding or feeding difficulties.

Endoscopy under general anaesthesia must be systematically performed in these severe forms to confirm the diagnosis and exclude an associated respiratory tract lesion, present in 18.9% of cases in the series published by Mancuso et al. [1]. Associated laryngotracheal lesions (laryngeal dyskinesia, vocal cord paralysis, subglottic stenosis, tracheomalacia) are more frequent in the pres-

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ence of severe laryngomalacia: Dickson et al. [2] reported associated lesions in 79% of cases (with subglottic stenosis in 73.3% of cases and tracheomalacia in 55.3% of cases) in infants with severe laryngomalacia versus 28.8% of infants with laryngomalacia associated with few signs of severity.

## Medical treatment and laryngomalacia

Due to the frequency and exacerbating role of pharyngolaryngeal reflux (PLR) in infants with laryngomalacia, anti-reflux treatment should be prescribed, despite the absence of evidence in favour of this approach in the literature (expert opinion).

Lifestyle and dietary measures must always be instituted (thickened milk, maintenance of posture after feeds, no bottle of water before lying down, raising of the head of the bed or mattress), and antacids in infants with regurgitation.

No studies have determined the optimal dose and duration of H<sub>2</sub> histamine antagonist or proton pump inhibitor (PPI) therapy or the preferred molecule. Ranitidine can be used at the dose of 3 mg/kg/day and PPIs can be used at the dose of 1 to 2 mg/kg/day. The efficacy of this treatment has been reported in several studies, but no double-blind trials have been conducted [3].

In 2011, the SFORL expert group recommended medical treatment for laryngomalacia with signs of severity, or in the presence of characteristic signs of pharyngolaryngeal reflux on pharyngolaryngeal endoscopy.

PPI therapy is also recommended postoperatively in infants treated by supraglottoplasty procedures: cases of postoperative stenosis have been reported in infants with gastro-oesophageal reflux, but the pathogenic role of gastro-oesophageal reflux simply remains suspected [4].

No study has confirmed the efficacy of local or systemic corticosteroid therapy in laryngomalacia.

## Surgical treatment of laryngomalacia

### Methods

Apart from tracheotomy, which remained the reference surgical treatment for severe forms of laryngomalacia for many years, several other surgical techniques have been proposed and have been gradually transformed into minimally invasive endoscopic approaches.

Variot [5], in 1898, was the first to propose resection of the excess mucosal tissue on the aryepiglottic folds, based on the post-mortem findings in a neonate with stridor. In 1922, Iglaue [5] was the first to perform partial epiglottectomy in a patient with laryngomalacia with a favourable outcome. In 1928, Hasslinger [5] performed endoscopic forceps division of the aryepiglottic folds in three patients with good results. In 1971, Fearon et al. [6] reported cases of suture of the epiglottis to the base of the tongue, allowing extubation of their patients. During the same period, cases of hyomandibulopexy were reported in France with satisfactory initial results [7], but this technique was subsequently abandoned. In 1981, Templer et al. [8] performed resection of the epiglottis, ventricular folds and aryepiglottic folds via lateral pharyngotomy in an 18-year-old patient with a satisfactory result.

The publication by Lane et al. [9] reporting endoscopic treatment of laryngomalacia with resection of excess supra-arytenoid mucosal tissue and the epiglottic mucosa using microinstruments (microforceps and microscissors) led to a renewed interest in these endoscopic treatments. One year later, Seid et al. [10] used the CO<sub>2</sub> laser to divide short aryepiglottic folds in three patients. Following numerous subsequent publications [11–13] endoscopic treatment of laryngomalacia has become the standard treatment.

### Anaesthesia

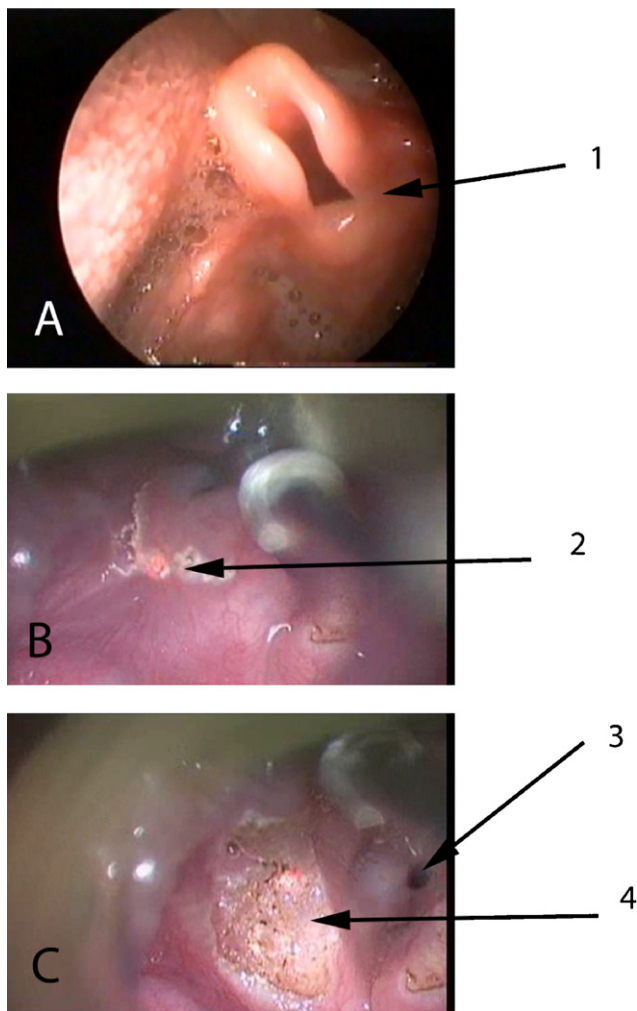
The main methods of ventilation are:

- mechanically controlled ventilation via a small calibre endotracheal tube is rarely used, as it interferes with the surgical procedure;
- spontaneous breathing anaesthesia, which constitutes the technique of choice of experienced anaesthetist-surgeon teams;
- intermittent apnoea technique that provides the surgeon with only a limited time to perform the procedure between two reintubations.

Jet ventilation, sometimes described for this surgery, has not been validated for laryngeal obstructive diseases such as severe laryngomalacia.

### Surgical technique

Most of the new surgical techniques used in this disease, called supraglottoplasties, are designed to reduce the excess tissues on supraglottic structures responsible for collapse. Several improvements have been proposed over time, not only concerning the endoscopic surgical technique, but also concerning the methods used to resect excess tissues. Since the first descriptions of supraglottoplasty [9,11–13], technical modifications have mainly concerned the site and extent of the tissues to be resected. The surgical technique [14] usually consists of division of short aryepiglottic folds, and sometimes a resection of excess supra-arytenoid mucosal tissue (Fig. 1), section of the median glossoepiglottic ligament with suspension of the epiglottis to the base of the tongue, partial epiglottectomy [15] or a combination of several of these techniques. Supraglottoplasty is usually bilateral, but some authors have highlighted the advantages and disadvantages of unilateral versus bilateral supraglottoplasty and have reported a lower risk of supraglottic stenosis after unilateral supraglottoplasty [16]. Various methods have also been proposed for resection of excess tissues: cold microinstruments (microscissors), CO<sub>2</sub> laser, Thulium laser [17], diode laser, microdebriders [14] and no differences in terms of the results obtained with these various techniques have been reported in the literature [18]. According to some authors, the advantages of laser and microdebrider compared to microscissors are the absence of intraoperative bleeding, a decreased risk of oedema and simplification of the operative technique. Lasers and microdebriders are more expensive than classical microsurgery instruments and lasers require special precautions to avoid the risk of fire during surgery.



**Figure 1** CO<sub>2</sub> laser division of the aryepiglottic folds in a non-intubated infant. A. Supraglottic stenosis with a tube-shaped epiglottis and short aryepiglottic folds (1). B. Start of CO<sub>2</sub> laser division of the aryepiglottic folds (2) at the free edge of the epiglottis and the ventricular fold. C. Appearance of the left aryepiglottic fold after resection (4) – glottis (3).

Hospitalisation is usually scheduled when upper airway endoscopy is indicated in the setting of severe laryngomalacia. Depending on the severity of the symptoms (especially respiratory distress) and the comorbidity associated with laryngomalacia, a bed is reserved in the paediatric intensive care unit postoperatively. Endoscopy under general anaesthesia may start with nasal endoscopy on induction of anaesthesia to allow dynamic examination of the infant's larynx under optimal conditions (sedated infant) and to detect any sleep-related or sleep-exacerbated diseases. Deeper general anaesthesia is then ensured by intravenous anaesthetics and anaesthetic gases and rigid endoscopy is ideally performed under spontaneous breathing. A dose of systemic corticosteroids (methylprednisolone 2 mg/kg) is administered at the beginning of the procedure. Local anaesthesia of the glottic and supraglottic regions is performed. Direct laryngoscopy (Pearson or Benjamin spatula) and rigid bronchoscopy are performed to confirm laryngomalacia and to define the most appropriate surgical

procedure based on the anatomical type of laryngomalacia, and to detect any concomitant respiratory tract lesions. Surgical treatment of laryngomalacia is then performed during direct laryngoscopy (suspension laryngoscopy) using an endoscope or an operating microscope. The surgical procedure most commonly performed is division of the aryepiglottic folds, possibly combined with resection of excess supra-arytenoid mucosa (Fig. 1). Resection of mucosal tissue must be performed very cautiously and must remain away from the midline in the inter-arytenoid zone to decrease the risk of postoperative supraglottic stenosis of the larynx. Laser can be used in continuous mode at a power of about 1 Watt for CO<sub>2</sub> laser or 1.5 Watt for Thulium laser. When using a microdebrider, the recommended power is 800 r.p.m. in oscillating mode, without irrigation, using a 2.9 or 3.5 mm diameter laryngeal blade. Haemostasis and cleaning of the operative field are performed with compresses soaked in adrenaline saline (1 mg of adrenaline in 10 ml of saline). Epiglottopexy is not performed as first-line procedure, except for a few teams [19,20], but only following failure of previous techniques with persistent posterior prolapse of the epiglottis. In this situation, laser vaporisation is performed on the lingual surface of the epiglottis over the median glossoepiglottic ligament to create a raw surface and epiglottopexy is then performed between the epiglottis and the base of the tongue with resorbable suture material (Vicryl 3/0). Finally, partial resection of the suprahyoid portion of the epiglottis (traction with forceps applied to the median aspect of the epiglottis and partial C-shaped epiglottectomy using microscissors or laser) is rarely performed and tracheotomy can be proposed following failure of these various procedures.

### Indications for surgical management

Laryngomalacia is the most common laryngeal abnormality and usually has a favourable outcome [6,21]. Only 10 to 20% of infants present severe laryngomalacia requiring surgical management [22,23]. The criteria used to define the severity of laryngomalacia vary from one author to another [5], but all authors take into account the severity of the symptoms associated with stridor. Surgery should therefore be proposed when laryngomalacia is associated with one or more of the following symptoms:

- dyspnoea with permanent and severe intercostal or xiphoid retraction;
- episodes of respiratory distress;
- obstructive sleep apnoea;
- episodes of suffocation while feeding or feeding difficulties;
- poor weight gain.

Note that some of these symptoms can be associated with other aetiologies, such as gastro-oesophageal reflux; which must be identified and treated before deciding to perform surgery. Finally, the family setting must also be taken into account in the treatment decision.

## Evaluation of surgical treatment – Efficacy

Surgical treatment of laryngomalacia provides rapid and lasting improvement in 70% to 100% of cases (Table 1). However, the results of the various published series must be compared very cautiously, as patient selection or postoperative evaluation of operated patients are often based on subjective criteria with marked variability between teams. A marked reduction or even complete resolution of stridor is usually observed after surgery, as well as resolution of the cyanotic and apnoeic episodes described by the parents and improvement of oxygen saturation on monitoring. After the first early postoperative phase (seven to 15 days), during which feeding can be difficult (especially after the use of laser techniques), a marked improvement of feeding is observed (absence of suffocation and rapid ingestion of bottles [less than 15 minutes]). However, deterioration may be observed in the presence of pre-existing swallowing disorders. Weight gain returns to normal after the first postoperative month. Improvement of respiratory difficulties is also observed, with resolution of intercostal recession. Some authors perform early follow-up endoscopy, before discharge from hospital, to eliminate any early synechiae. All authors perform nasal endoscopy at an office visit, three to four weeks after surgery. Nasal endoscopy reveals good healing of the larynx and marked improvement of collapse of the supraglottic part of the larynx.

Evaluation of efficacy should be based on objective clinical criteria (height and weight gain) or functional criteria (polysomnography, echocardiography in the presence of pulmonary artery hypertension). Polysomnography allows recording of apnoeas, hypopnoeas, and oxygen saturation and can be combined with measurement of gas exchanges during sleep especially transcutaneous CO<sub>2</sub> (PtcCO<sub>2</sub>). Several teams propose polysomnography before

any surgical procedure [24,25], except in an emergency. This initial examination can then be used as the reference baseline examination to be compared with postoperative recordings in the event of failure of treatment or only partial clinical improvement.

Several series have reported the objective results of the efficacy of surgical treatment of laryngomalacia based on preoperative and postoperative polysomnography [24–27] with improvement of the apnoea/hypopnoea index (AHI). However, these series were based on limited sample sizes and postoperative polysomnography was performed at variable intervals after surgery. Polysomnography clearly constitutes a very useful tool for evaluation of the severity of laryngomalacia and the efficacy of treatment. However, routine use of polysomnography would be difficult in most centres due to the prohibitive cost and especially the limited access to this examination with long waiting times.

Other authors have reported improvement of gastro-oesophageal reflux [28] after aryepiglottoplasty.

## Risks of failure of surgery

The risk of treatment failure is higher in infants with a concomitant disease. Denoyelle et al., based on a series of 136 infants, observed failure of surgical treatment ( $n=5$ ) (or only partial improvement of the symptoms,  $n=7$ ) only in patients presenting laryngomalacia associated with other congenital anomalies [4]. Schroeder et al. also had to more frequently use postoperative adjuvant respiratory therapy (nebulization, noninvasive ventilation, oxygen therapy, intubation) in children with encephalopathy or laryngomalacia associated with subglottic stenosis [29]. Valera et al. observed treatment failure in three patients with associated tracheomalacia or encephalopathy [27] in a

**Table 1** Various series published in the literature according to [18].

	O'Donnel et al. [18]	Martin et al. [43]	Toynton et al. [23]	Roger et al. [22]	Polonovski et al. [31]	Remacle et al. [44]	McClurg et al. [42]	Whymark et al. [20]
Year	2007	2005	2001	1995	1990	1996	1994	2006
Number of infants	84	30	100	115	39	12	24	59
Improvement of stridor (% of cases)	88.1	90	86	89	97	100	71	73
Micro-inhalations or aspirations (% of cases)	7.1	13	6		0	0		7
Need for tracheotomy (% of cases)	0	6.7	1	1.7	2.6			14
Need for redo procedure (% of cases)	4.8	3.3		1.7	5.1		50	12
Mean hospital stay	1.5 day (13.2 in the presence of comor- bidity)		5.3 day (12.9 in the presence of comor- bidity)		48 h for most infants			



series of 59 infants treated surgically for severe laryngomalacia, while no failure was observed in infants with isolated laryngomalacia. In contrast, out of ten infants with a neurological condition, one required redo laryngoplasty and six required tracheotomy after failure of supraglottoplasty. In the same series, ten infants presented cardiac anomalies and three of them required tracheotomy after failure of surgery. Two of the eight infants with congenital malformation syndrome (one infant with Pierre Robin sequence and another with Down syndrome) required tracheotomy. In the absence of comorbidity, age at the time of surgery less than 2 months appears to be associated with a higher incidence of redo supraglottoplasty compared to children operated after the age of 2 months (6.4% vs 5.3% in the study by Hoff [30]).

### Evaluation of the surgical treatment – Complications

Complications of surgical treatment of laryngomalacia are fairly rare in view of the young age of the infants concerned and the frequency of comorbid conditions, particularly congenital malformations. Most series [3,4,31] report complication rates less than 10%. However, the presence of neurological or cardiac comorbidity and age less than 2 months constitute major risk factors for failure and/or complications [30]. Laryngeal spasm or laryngeal oedema during the perioperative period may require intubation.

Local postoperative complications include:

- granulomas, haemorrhage, adhesions that can cause supraglottic stenosis, infection, transient aspiration. An overly aggressive surgical technique (especially laser or excessive resection with cold instruments) or gastro-oesophageal reflux could explain these complications, but no particular technique and poorly controlled gastro-oesophageal reflux do not appear to be associated with a higher risk of complications [32];
- failure to recognize an associated laryngotracheal lesion (subglottic stenosis, tracheomalacia) that may compromise postoperative extubation, requiring tracheotomy [30].

Long-term complications are essentially lower respiratory tract infections related to aspiration or complications related to decompensation of associated malformations (cardiac, renal, cerebral etc.).

### Methods and indications for noninvasive ventilation in laryngomalacia

#### Indications

Noninvasive ventilation (NIV) decreases the respiratory work of infants with upper airway obstruction associated with alveolar hypoventilation [33,34], especially laryngomalacia [35,36]. It allows improvement of nocturnal gas exchanges [36] and maintenance of satisfactory weight and height gain [36]. Fauroux et al. demonstrated the efficacy of Bilevel Positive Airway Pressure (BIPAP) comprising pressure support and positive end-expiratory pressure [36] in 12 children

with a mean age of 33 months (8–79 months, three infants less than 1 year old). Essouri et al. also demonstrated the efficacy of continuous positive airway pressure (CPAP) ventilation [35] in ten infants with a mean age of 10 months (3–18 months) presenting upper airway obstruction (five cases of laryngomalacia, three cases of tracheomalacia).

NIV can avoid tracheotomy and its consequences: canula obstruction, discomfort, delayed speech development, family consequences [37,38]. In 2006, a consensus (SFAR, SPLF, SRLF) considered that NIV should be proposed for the management of acute respiratory failure secondary to laryngo-tracheomalacia [39]. However, no guidelines are available for isolated laryngomalacia, either in a context of acute decompensation or for chronic management.

The expert group proposed the use of NIV in laryngomalacia, either isolated or part of a congenital malformation syndrome, with signs of severity [35,36] complicated by sleep-disordered breathing (apnoeas, gas exchange anomalies), poor weight gain or, in extreme cases, pulmonary artery hypertension when surgical treatment cannot be proposed (or while waiting for surgery) or when medical and surgical treatment is not sufficiently effective.

#### Methods

No guidelines are available concerning the modalities of NIV for infants with laryngomalacia. However, the guidelines published by the *Association Française contre les Myopathies* and the *Haute Autorité de la santé* (French National Authority for Health) for NIV in children with neuromuscular diseases (*Modalités pratiques de la VNI en pression positive, au long cours, à domicile, dans les maladies neuromusculaires*) can be used as a guide. NIV must be performed by an experienced team as part of multidisciplinary management (ENT and maxillofacial surgeons, paediatric respiratory physicians, nurses, social workers). It can only be performed with the family's agreement and active cooperation (HAS recommendation concerning NIV in children with neuromuscular diseases).

CPAP or BIPAP can be used, always with a leakage system. The sensitivity of the inspiratory trigger of the ventilators (BIPAP mode) available in France is usually poorly adapted to infants [35,40]. It is therefore preferable to start with CPAP ventilation, which maintains constant upper airway opening throughout the respiratory cycle [33–35], as Essouri et al. showed that BIPAP causes more marked asynchronism than CPAP. However, BIPAP with a minimum frequency adjusted to the respiratory rate in infants (thereby avoiding the use of the inspiratory trigger) can be used to limit this asynchronism. The pressures used depend on the course of clinical (clinical examination, weight gain, the infant's development) and functional parameters (apnoeas, gas exchanges).

The interface used must be adapted to the infant's face. A nasal mask is generally used and the choice of this mask is essential, either a commercially available mask (but few masks are available for low weight infants) or handmade mask moulded onto the infant's face. The mask must be comfortable, must not irritate the skin, and must not cause any leaks. Use of a teat, to limit mouth leaks, is often effective and usually better tolerated than a chinstrap. Assessment of the short-term tolerability of the mask is an essential

part of surveillance. NIV is generally used during sleep, which can represent the majority of the 24-hour period in infants. There is a risk of skin lesions or facial deformities secondary to the pressures exerted by the mask on growing facial structures [41]. In a cohort study of 40 ventilated patients (16 infants with OSAS, 14 infants with neuromuscular disease, ten infants with cystic fibrosis) with a mean age of 10 years (0.6–18), Fauroux et al. reported skin lesions in 48% of cases, maxillary retrusion in 37% of cases, and global flattening of the face in 68% of cases. No correlation was observed between these complications and the infant's age, the type of mask, or the underlying disease. In contrast, the risk increased with the duration of use of daytime NIV (OR = 6.3). Follow-up by a maxillofacial surgical team is therefore essential.

## Conclusion

Laryngomalacia is the most common laryngeal disease in infants. It is poorly tolerated in 10% of cases, requiring assessment and surgical management as well as management of any associated gastro-oesophageal reflux. Surgery effectively controls symptoms, but few data are available concerning objective assessment of the results: preoperative and postoperative objective assessment of these infants must be performed whenever possible. However, NIV may be indicated in some infants with comorbid diseases or who fail to respond to surgical treatment.

## Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

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